

Case Report

Biliscrotum and Retroperitoneal Biloma: Spontaneous Rupture of the Biliary System presenting as an Incarcerated Inguinal Hernia

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Accepted 5 October 2005

ABSTRACT

Spontaneous rupture of the biliary system is a well documented condition in infants¹ but is rare in adults.² We report the case of a 73-year-old gentleman who presented with clinical signs and symptoms mimicking that of a strangulated right inguinal hernia. At emergency operation the scrotum was found to contain bile. Following radiological imaging and exploratory surgery, a large retroperitoneal biloma was found. We discuss the clinical signs associated with biliscrotum and retroperitoneal biloma and describe our operative management of this patient. We review the previously reported cases of these rare clinical entities. We found that our case exhibited similarities in terms of the age of presentation and presence of distal common bile duct stones. This is, to our knowledge, the only reported case of a patient presenting with biliscrotum secondary to the assumed spontaneous rupture of the common bile duct and development of a retroperitoneal biloma.

CASE REPORT A 73 year old male presented as an emergency with sudden onset of severe pain distributed in the right inguinal region, anorexia and vomiting. The patient had no pyrexia and was haemodynamically stable.

Examination revealed a mass within the right inguinal region, a fullness in the right iliac fossa and tender enlargement of the right side of the scrotum. These signs were associated with a brown discoloration of the skin overlying the right inguinal mass and the right side of the scrotum. His history included a right inguinal hernia repair in 1973.

Blood results on admission revealed obstructive liver function tests. The patient proceeded to emergency contrast-enhanced CT-scan of abdomen. A right inguinal hernia containing mesenteric fat and vessels was reported, with a fluid collection extending along the iliopsoas muscle and around Gerota's fascia of the right kidney. Calculi were noted within the gallbladder.

The patient proceeded to emergency right inguinal hernia exploration. A process of mesenteric fat was found within the inguinal canal. In addition, there was a copious amount of green fluid that tracked along the mesenteric fat process and pooled in the right hemiscrotum (*Fig 1*). An oedematous and inflamed spermatic cord was found. The patient underwent repair of the inguinal defect.

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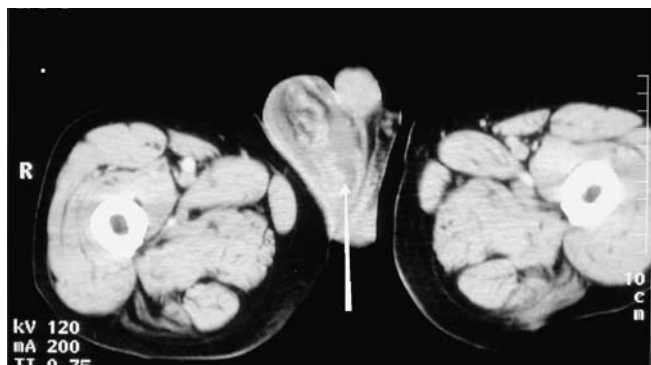


Fig 1. Biliscrotum. Axial CT scan at the level of the scrotum/inguinal canal showing bile extending around the pampiniform plexus.

The patient's clinical status improved post-operatively. His obstructive liver function tests and bilirubin levels normalised. A CT scan was repeated on post-operative day 3 in order to assess the size of the retroperitoneal collection and to guide towards aetiology. This described an increase in the size of the retroperitoneal fluid collection since the previous scan. Calculi were still noted within the common bile duct (CBD) which was 1.2 cm in diameter. (*Fig 2*).

The patient proceeded to exploratory laparotomy. Following extensive examination of the gallbladder, bile ducts, duodenum, liver and colon, there was no obvious perforation, leak of bile or connection with the retroperitoneum. There was no bile collection found intra-peritoneally. Examination of the gallbladder revealed multiple gallstones and exploration of the CBD was performed. A biliary fogarty catheter trawl of the CBD resulted in the retrieval of 2 large stones. An intra-operative cholangiogram revealed no perforation or abnormalities of the CBD apart from distension. A t-tube drain was placed within the CBD and a cholecystectomy was performed. The right retroperitoneum was entered lateral to the ascending colon. A large collection of bile stained liquid was encountered and evacuated. Drains were placed in the right retroperitoneum and around the gallbladder bed.

The patient's post-operative course was uncomplicated. The drainage from the retroperitoneal drain progressively decreased and there was consistent bile drainage from the t-tube. A t-tube cholangiogram on day 10 demonstrated no residual stones stricture or perforation. Pathological examination of the gallbladder post-operatively revealed no perforation or tissue necrosis and

confirmed the finding of large gallstones intraluminally.

Following transfer for further rehabilitation, the patient was discharged from hospital without complication and at 1 year post-operatively is continuing to progress well.

DISCUSSION

The presence of a bile collection within the retroperitoneum is extremely rare.³ Since 1975 only 5 other cases have been reported.³⁻⁷ Two describe complications arising from invasive surgical procedures^{4,5} and 3 were attributable to the spontaneous rupture of either the gallbladder⁶ or CBD.^{3,7}

The authors have assumed the site of perforation was within the CBD given the dilatation of the duct at the time of the surgery and the obstructive liver function tests. In addition, the presence of CBD intra-ductal stones, an intra-operatively normal duodenum and the post-operative gallbladder pathology findings which demonstrated no fistulae or perforation support our assumption. We postulate that the release of pressure on the biliary system following perforation had allowed the resolution of the liver function tests and that the subsequent absence of extra-ductal contrast on cholangiogram could have resulted from a resealed perforation following decompression of the obstructed system. This precedent has been noted to occur in previous cases.⁷

Previous authors have either surgically or pathologically been able to identify the cause of

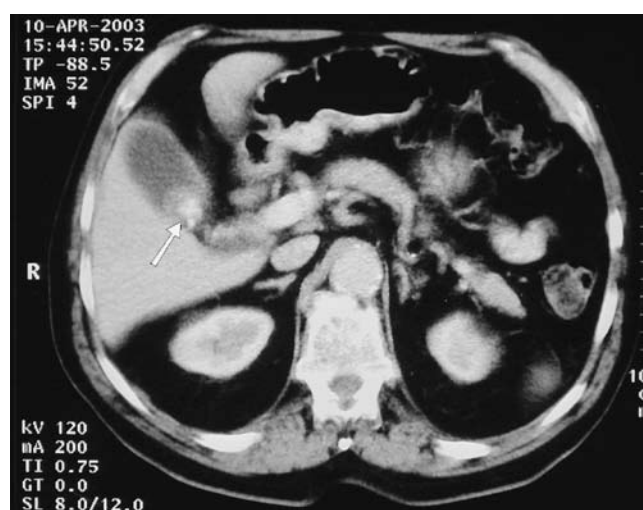


Fig 2a Axial CT-scan at the level of the gallbladder, showing a large calculus within the gallbladder and a further small calculus in adjacent dilated CBD.

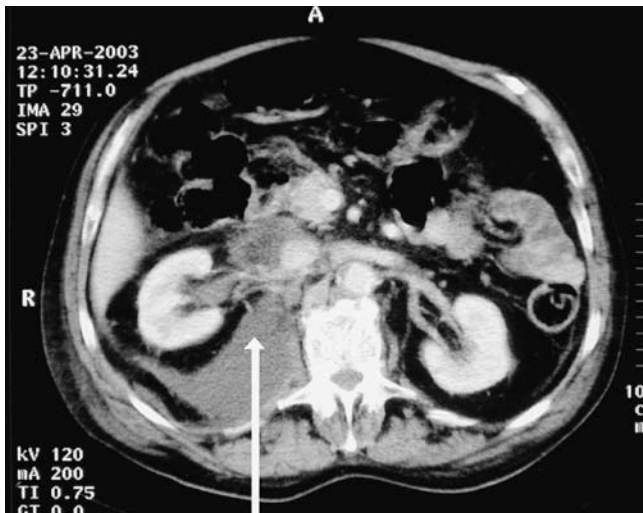


Fig 2b. Bile is extravasating from the biliary tree, around the inferior vena cava and right kidney.

the retroperitoneal connection. Such authors have described internal biliary fistula (secondary to postoperative CBD stricture), or gallbladder fistula to the retroperitoneum.⁶ Satake³ described the first retroperitoneal biloma secondary to spontaneous perforation of the CBD due to an erosive obstructing CBD stone. Hsieh,⁷ in comparison with our case, described a 74-year-old male with a spontaneous retroperitoneal biloma. Following percutaneous drainage of the biloma and the gallbladder, cholangiography demonstrated a calculus within the distal CBD, but as no other abnormality was found, the site of perforation, was assumed to have sealed spontaneously.

Whipple⁸ first described the term biloma in relation to a cystic swelling containing bile stained fluid. Gould and Patel⁹ further refined this definition as an encapsulated bile collection outside the biliary tree, which was again expanded to include intra-hepatic collections.¹⁰ Colovic⁴ was the first author to use the term retroperitoneal biloma to describe a retroperitoneal collection of bile. Hsieh⁷ continued this precedent even in the absence of a demonstrable capsule on CT scan. Vasquez¹¹ *et al* in their review of 21 bilomas found that “on CT scans, bilomas do not often show an identifiable capsule”.

Previous authors have reported the clinical signs in association with these conditions. Neoptolomos⁵ described an area of “discolouration in the right flank” and right sided scrotal swelling which “contained fluid which proved to be bile on aspiration (Biliscrotum)”. The formation of Biliscrotum relies on bile tracking from the retroperitoneum along fascial planes formed by

embryological fascial planes. Horowitz⁶ details a palpable mass within the right inguinal region, this patient also underwent an operation for incarcerated inguinal hernia. Satake³ and Colovic⁴ both described a large mass felt in the right lower abdomen in their reports.

This report emphasises the importance of being aware of the clinical signs associated with retroperitoneal biloma and involvement of distal CBD stones or operative trauma in the aetiology of this condition.

CONFLICT OF INTEREST

None Declared

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